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Case report

Long-term use oral bisphosphonate-related osteonecrosis of the jaw without dental extraction in elderly: A case report

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ABSTRACT

Bisphosphonate-associated osteonecrosis of the jaws (BRONJ) may have significant morbidities ranging from minimal discomfort to significant loss of function. We report a case of jaw osteonecrosis resulting from long-term use of bisphosphonate. An 83-year-old female, a long-term user of alendronate, developed jaw osteonecrosis without a history of recent dental extraction. Facial bone computed tomography of the patient showed a destructive osteolytic change of the whole right mandible bone with adjacent soft tissue swelling. After surgical debridement, antibiotic treatment, and discontinuation of alendronate, the patient recovered very well. Although uncommon, BRONJ has gained extensive attentions. Physicians should be aware of the possibility of BRONJ among long-term bisphosphonate users, and a collaboration of clinical pharmacist may be helpful in early detection.

1. Introduction

Osteoporosis is common in the elderly population and a significant burden in terms of impaired quality of life.[1] It is estimated that 10 million people aged above 50 years in the United States have osteoporosis.[2] Oral or intravenous bisphosphonates are common pharmacologic agents to treat osteoporosis. To date, more than 190 million prescriptions for oral bisphosphonates have been dispensed worldwide,[3] and long-acting bisphosphonates have become the mainstream of osteoporosis treatment.[4,5] In addition to the antiresorptive effect of bisphosphonate, the potential of osteoclast-mediated bone resorption in treating other diseases has gained more and more attention. Although bisphosphonate is generally agreed to be safe, increasing body of evidences suggest that long-term bisphosphonate use may be associated with an unusual condition—jaw osteonecrosis.[6,7] Herein, we report a case of long-term bisphosphonate user who developed unexplained jaw osteonecrosis and to evaluate the treatment response.

2. Case report

An 81-year-old lady who had history of bronchial asthma, hypertension, gastric ulcer, and osteoporosis took alendronate (70 mg/wk) for two years in a community hospital in Taiwan. However, she felt persistent pain in the right cheek for the past six months, so she visited geriatric outpatient department for this condition and polypharmacy. The patient stated that she had persistent right cheek pain for six months and the pain still remained despite a visit to a dentist. She was suggested to visit a tertiary medical center for further opinion and treatment. However, the development of gastric ulcer delayed her visit for the pain of her right cheek. The pain deteriorated progressively, which caused her difficulties in

Fig. 1. Raw bone exposure at lower right mandible area with purulent pus discharge, about 3.0 × 1.5 cm.

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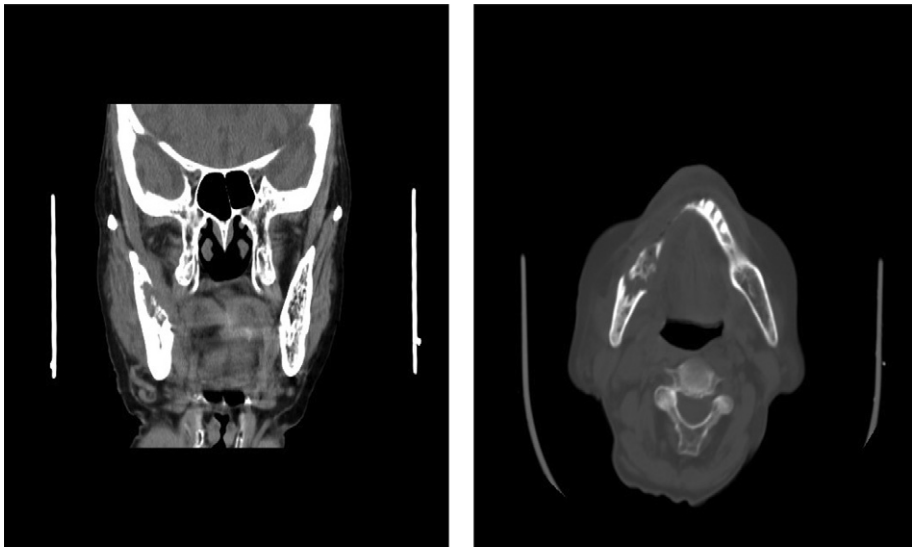

Fig. 2. Destructive osteolytic change in the whole right mandible bone with adjacent soft tissue swelling.

eating. Therefore, she visited geriatric outpatient department because of this aggravated cheek pain and the burden of polypharmacy. Physical examinations revealed painful swelling of the right cheek with limited mouth opening, bone exposure at the lower right area with purulent discharge (about 3.0 × 1.5 cm in size and #27 elongation ulcer area) (Fig. 1). Dental panoramic radiography was done showing a diffuse osteolysis of the right mandibular body and a sequestrum near the alveolar crest. The clinical impression was jaw osteonecrosis with secondary infection, which may be associated with long-term use of bisphosphonate. Facial bone computed tomography was done revealing a destructive osteolytic change in the whole right mandible bone with adjacent soft tissue swelling (Fig. 2). Throughout the whole course of treatment, there was neither trauma nor denture implant nor previous dental surgery, so the patient was admitted under impression of bisphosphonate-associated osteonecrosis of the jaws (BRONJ) with secondary infection. During her hospital admission, surgical debridement, antibiotic use, and discontinuation of alendronate were done, and the patient recovered well from the BRONJ.

3. Discussion

Bisphosphonates are potent inhibitors of osteoclasts, which may unavoidably retard the skeletal repair processes associated with trauma. It has been hypothesized that the constant use of jaw bones may result in minor traumas, and the associated continuing bone remodeling is the cause of a preferential accumulation of bisphosphonates and leading to osteonecrosis.[8,9] The incidence of BRONJ in long-term bisphosphonate users ranged from 1%–10%.[10–13] Because osteoporosis is an age-related disease, the prevalence and incidence rise with older age. Therefore, long-term use of bisphosphonate may be a common phenomenon in the aging population. Two theories have been proposed to clarify the pathogenesis of BRONJ. One focused on the bisphosphonate-induced osteoclast inhibition and the other favors the antiangiogenic mechanisms to be the contributing factor.[8] In most cases, the development of osteonecrosis of the jaws among those long-term bisphosphonate users is associated with traumas, predominantly dental extraction.[14–17] However, even rarer, some patients may spontaneously develop BRONJ in the absence of overt trauma or dental extraction.[18]

The American Association of Oral and Maxillofacial Surgeons has published the BRONJ diagnosis criteria in 2007,[3] and this reporting case was present with all three diagnostic elements of BRONJ, i.e., (1) exposed bone being present for longer than eight weeks, (2) exposure to bisphosphonates, and (3) no history of radiation therapy to the maxilla or mandible. Although BRONJ is rare and the BRONJ without preceding trauma is rarer, physicians treating osteoporosis should bear in mind of this uncommon complication. However, balancing the risks and benefits of bisphosphonate use, bisphosphonates are still considered safe in clinical practice. The potential fracture risk secondary to the discontinuation of bisphosphonates should be balanced by other agents or treatment strategies. Collaboration with pharmacists in clinical practice may be important to gain better insights into the optimal treatment of patients with osteonecrosis of the jaws and to promote early detection of BRONJ.

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